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Painful Proximal Oriented Large Heterotopic Spur Formation In An Active Adult Non-Traumatic Amputee

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Abstract

Heterotopic ossification (HO) is excess bone growth in soft tissues, typically juxta-articular and interfascicular, with varying incidence. HO has been well documented in traumatic amputees but less frequently observed in the non-traumatic amputee population. Symptomatic HO usually includes pain during prosthetic use, with management involving prosthetic adjustments for comfort. This atypical case highlights a non-traumatic amputee developing proximal oriented large spur formation that was painful not with ambulation but with doffing of his prosthesis.

Case Description

A 58 year old gentleman with a history of atrial fibrillation on coumadin and peripheral vascular disease presented to an emergency room in Italy in June 2015 with shortness of breath. The patient was in acute respiratory failure requiring intubation and mechanical ventilation and prolonged vasopressor support for septic shock. He required 15 days of extracorporeal membrane oxygenation for support as his respiratory function improved with antibiotics. After weaning off vasopressors, both lower extremities were found to be hypoperfused, requiring a right transtibial amputation and a left transfemoral amputation. The left lower extremity required an additional debridement for surgical site infection and a prolonged antibiotics course. He then returned to the United States in September 2015.

The patient had inpatient pre-prosthetic training in September 2015 and inpatient prosthetic training in February 2016, demonstrating proficiency and independence in ambulation and activities of daily living with his prostheses. At that time, he did not have any atypical pain, nor was there any evidence of heterotopic ossification.

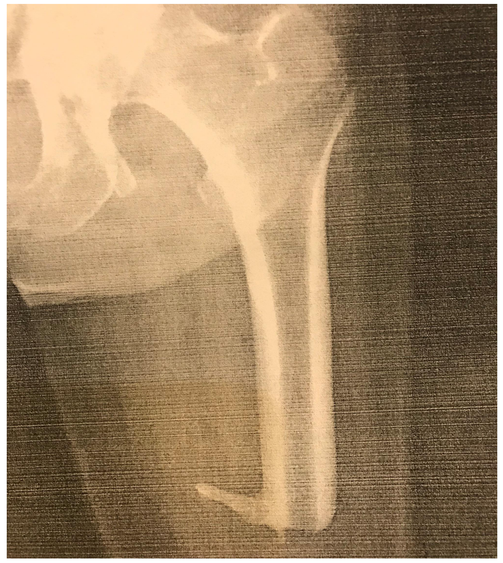
The patient was highly active with his tight fitting prostheses. However, he began to complain of severe pain in the left residual limb, not with weight bearing, but only when he removed his residual limb from his socket. Upon his follow up appointment with his physiatrist, an X-ray was ordered which revealed a 3.9 x 2.3cm calcified lesion at the medial aspect of the femoral shaft of his residual left femur. This heterotopic spur was oriented proximally. He was referred to plastic surgery, who removed the spur and noted that the spur had a significant blood supply. Post-operatively, the patient was pain free and continued to be active with his prostheses.

Heterotopic Ossification

Heterotopic ossification (HO) is excess bone growth in soft tissues. Typically it develops outside the borders of normal periosteum and has its own vascular supply [1]. It can be contiguous with the skeleton, however, it does not involve the periosteum, and once matured, it contains cancellous and lamellar bone, bone marrow, and vessels [2]. While the pathogenesis is not completely understood, it is believed that dormant osteoprogenitor stem cells within soft tissue are triggered to differentiate into osteoblasts ultimately leading to bone formation [3]. This bone growth may or may not be symptomatic. Symptoms include pain during prosthetic use [4]. Typically, symptoms can be alleviated through prosthetic adjustments for a more comfortable fit [4]. Heterotopic ossification can create a pressure-sensitive area requiring multiple modifications of the prosthesis [5]. Conservatively, nonsteroidal anti-inflammatory drugs and bisphosphonates are used to manage progression, along with therapy to preserve range of motion [3]. External beam radiation therapy is also available but at a more significant cost [3]. A minority of patients may require surgical excision of the ectopic bone to relieve discomfort, however, this requires mature bone [3]. Maturation of bone occurs approximately 12 to 18 months after injury [4]. During amputation, the surgeon should consider resecting the bone such that periosteum covers the distal end [5]. If not well covered with periosteum, spike-like heterotopic ossification might develop in the soft tissue causing pain and irritation [5].

Discussion

The pathophysiology of HO is not very well described in the literature. It is thought to be due to the transformation of dormant osteoprogenitor stem cells into osteoblasts, leading to bone formation [3]. Various humoral, neural, and local factors must come together to create the necessary environment for HO to occur [7]. When the periosteum covering bone that is retained is stripped, ectopic bone formation can occur [8]. This results in the formation of a simple bone spur. However, what makes this case unique is the extension of the bony growth into adjacent soft tissue as well as its irregular shape. An extensive literature review revealed only one case report of heterotopic ossification in the residual lower limb in an adult non-traumatic amputee [9]. To our knowledge, heterotopic ossification in an adult non-traumatic amputee is a rare occurrence. In addition, the patient's presentation of pain with doffing of the prosthesis, along with his proximal orientation of his heterotopic spur, appear to be unique. Dudek et al. [5] described two separate case reports of patients with HO that caused significant residual limb pain post-amputation. The first was a 39-yr-old woman who had a left transfemoral amputation after a traumatic event. Radiologic examination at 8 months post-amputation revealed a bone growth along the femoral shaft extending from the amputation site to adjacent soft tissues. Release of the sciatic nerve as well as adjustments to her prosthetic socket were not helpful in relieving the magnitude of her limb pain. Surgical excision was performed 18 after amputation with significant relief of her pain along with increased functional use of the prosthesis. The second was a 59-yr-old man who also underwent a left transfemoral amputation after a traumatic injury. His radiograph revealed bony spurring at the distal end of the femur as well as bony formation in adjacent soft tissues. The patient was limited in movement as weight-bearing on the amputated limb caused him pain. His pain resolved after a new prosthetic socket provided pressure relief at the distal lateral femur. Kömürcü et al. [10] described a 35-yr-old man who also had a transfemoral amputation following a traumatic injury. Sonographic as well as radiographic imaging of the right knee revealed a bony spur on the tibia. There was no mention of management of this patient's condition as the report only focused on diagnosis of the bony growth. Most recently, Melcer at al. [6] presented the occurrence of HO in a small series of combat amputees from Afghanistan and Iraq wars. Roughly one-third of patients with at least moderate HO showed no adverse symptoms. This finding highlights that not all HO cases are generators of pain and can be asymptomatic. Other cases showed that HO can affect pressure-sensitive areas of the amputated limb with adverse symptoms prompting management with prosthetic adjustment or even surgical excision. Further, one patient care issue they explored was the potential beneficial use of HO in prosthetic fitting in transfemoral amputees. When the bony outgrowth expands to surround the distal portion of the amputated bone, it becomes useful as an anatomical support structure that the prosthetist can use to improve the fitting of the residual limb into the prosthetic socket. We hypothesize the patient's large spur formation with characteristics of HO is related to his high activity in tight fitting prosthesis while on Coumadin, creating local trauma that would lead to hematoma and calcium deposition. Although it may be possible that bony overgrowth can occur in a skeletally mature patient, our radiologic findings point to HO as the pathology since the bony spike is not contiguous with the periosteum of the residual limb.



Radiograph of the patient's left residual limb in June 2017, 2 years after initial hospitalization.

Conclusion

Heterotopic ossification can be a pain generator in adult, non-traumatic amputees. Further, it remains to be determined whether the pathogenesis differs between traumatic and non-traumatic amputee populations.

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